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ACTIVITY AND PARTICIPATION IN PRESCHOOL CHILDREN WITH DIFFERENT INJURY TYPES OF OBSTETRIC BRACHIAL PLEXUS PARALYSIS

ORIGINAL ARTICLE

ABSTRACT

Purpose: Obstetric brachial plexus palsy (OBPP) causes various disorders in the musculoskeletal system. Studies investigating activity and participation in different types of injury are limited. This study aimed to determine the activity and participation levels of preschool children with different injury types of OBPP.

Methods: The 112 children with OBPP, aged between 44 and 77 months, were included. The children were grouped according to the Narakas Classification System: Type 1 (n=7), Type 2 (n=91), Type 3 (n=2), and Type 4 (n=12). Both Type 3 and Type 4 represented a total brachial plexus injury. The Pediatric Outcome Data Collection Instrument (PODCI) was used to evaluate activity and participation.

Results: Narakas Type 1 and Type 2 had similarly high scores in PODCI Global (p<0.001), and PODCI Upper Extremity (p<0.001), Pain/Comfort (p<0.001), and Sport (p<0.001) subscales as compared with Narakas Type 3/4. In the PODCI Happiness subscale, Narakas Type 1 had the highest score, followed by lower scores of Narakas Type 2 and Type 3/4 (p<0.001).

Conclusion: Preschool children with total OBPP had lower levels of activity and participation, less use of their arm in daily living activities, and express a lower level of sport participation compared to those with upper plexus injuries (Narakas Type 1 and Type 2). As activity and participation problems persist or even got worse as the children grow, it is essential to evaluate both activity and participation, particularly in those with total plexus injuries, in preschool age and preferably earlier.

Key Words: Activities of Daily Living; Birth Injury; Brachial Plexus; Health Status; Participation.

OBSTETRİK BRAKİAL PLEKSUS PARALİZİSİNDE FARKLI YARALANMA TİPLERİNE SAHİP OLAN OKUL ÖNCESİ YAŞTAKİ ÇOCUKLARDA AKTİVİTE VE KATILIM

ARAŞTIRMA MAKALESİ

ÖZ

Amaç: Obstetrik brakial pleksus palsi (OBPP), kas-iskelet sisteminde çeşitli bozukluklara yol açmaktadır. Aktivite ve katılım alanında çok az sayıda çalışma bulunmaktadır. Bu çalışmada, okul öncesi yaşta OBPP'nin farklı yaralanma tiplerinin aktivite ve katılım düzeylerinin belirlenmesi amaçlandı.

Yöntem: Çalışmaya 44-77 aylık OBPP olan 112 çocuk dahil edildi. Çocuklar Narakas Sınıflamasına göre gruplandırıldı: Narakas Tip 1 (n=7), Tip 2 (n=91), total brakial pleksus yaralanmasını içeren Narakas Tip 3 (n=2) ve Tip 4'ü (n=12). Çocukların aktivite ve katılım alanları Pediatrik Sonuç Veri Toplama Aracı (PODCI) kullanılarak değerlendirildi.

Sonuçlar: PODCI Global (p<0,001), PODCI Üst Ekstremitte Fonksiyonları (p<0.001), Ağrı (p<0,001), Fiziksel Fonksiyon ve Spor (p<0,001) skalalarında Narakas Tip 1 ve Tip 2 içerisinde yer alan çocukların benzer ve yüksek skorlar aldığını görüldü. Bununla birlikte bu iki grubun sonuçları Tip 3/4'ten daha yüksekti (p<0,05). PODCI'nin Mutluluk/Memnuniyet alt ölçeklerinde Tip 1 en yüksek puana sahipken, Tip 2 ve Tip 3/4 daha düşük puanlara sahipti (p<0,001).

Tartışma: Okul öncesi yaş grubunda total brakial pleksus hasarına sahip olan çocukların aktivite ve katılım seviyelerinin, günlük yaşam aktivitelerinde üst ekstremitte kullanımlarının ve spora katılmalarının üst brakial pleksus yaralanması olan Narakas Tip 1 ve Tip 2 yaralanması olan çocuklardan düşük olduğu görüldü. Aktivite ve katılım problemlerinin okul öncesi yaştan sonra aynı şekilde devam ettiği veya daha belirgin hale geldiği bildirildiğinden, özellikle total pleksus yaralanmalı çocukların tıbbi takiplerinde okul öncesi yaş ve daha öncesinde aktivite ve katılım değerlendirmeleri yapılarak tedavilerinde bu alanlara önem verilmelidir.

Anahtar Kelimeler: Günlük Yaşam Aktiviteleri; Doğum Yaralanması; Brakial Pleksus; Sağlık Durumu; Katılım.

INTRODUCTION

Obstetric brachial plexus palsy (OBPP) is a flaccid paralysis that occurs as a result of injury to brachial plexus (BP) during delivery (1). The incidence of OBPP ranges from 0.42 to 5.1 per thousand births. The extent of the damage may vary depending on the severity of the injury: from single nerve root to total root lesions. The clinical presentation of OBPP is a broad spectrum from transient functional insufficiencies to lifelong total disabilities, depending on the severity of the injury. Regular physiotherapy sessions and primary (and secondary) surgeries aim to minimize the disorders in the body structures and functions and enhance the patient's independence in activities of daily living (2-4).

The BP injury could lead to various disorders in body structures and functions; including denervation-related muscle weakness, agonist-antagonist muscle imbalance, muscle shortness, joint contractures and/or instability (3,5), sensory problems (6,7), spinal problems (8), impaired automatic arm movement (9), and deficits in normal motor development (10). In addition to body structure dysfunctions, activity and participation restrictions are also common (11). However, three different reviews investigating clinical evaluations and outcome measures in OBPP reported that the studies merely focused on body structure/function and that more studies are needed to investigate the domains of activity, participation, environmental, and personal factors according to the International Classification of Functioning, Disability and Health (ICF) (12-14).

There are a limited number of studies investigating activity and participation in children with OBPP; however, in these studies, children with different types of BP injuries were included in a single group and compared with normative values of healthy children (15-17). Nevertheless, OBPP includes a wide range of functional variations depending on the extent of nerve injury at birth. It has been reported that while children with upper truncus injuries gain up to 90-95% functional recovery during the first years of life, those with more severe nerve injuries may suffer permanent disorders (2,3,18,19). Despite this diversity, the differences in activity and participation levels of children with different types of BP injuries have not been investigated.

Despite several studies investigating body structure/function, there is minimal information on activity and participation levels of children with OBPP with different injury types (12-14). Therefore, the present study aims to investigate the activity and participation levels of preschool children with different types of OBPP.

METHODS

This cross-sectional study was conducted at Hacettepe University between November 2018 and November 2019, and approved by Hacettepe University Non-Interventional Clinical Researches Ethics Board (Approval Date: 06.11.2018 and Approval Number G0 18/1020-32). The families were informed about the study and those willing to participate in signed written consent forms. Medical records of all patients were examined, and Narakas classifications were determined by the medical doctor (AU) and referred to the physiotherapy clinic. All evaluations were performed by the same physiotherapist (KD) had eight years of experience in the field of pediatric hand rehabilitation and OBPP and who was blind to the Narakas Classification of the children. The inclusion criteria were being in preschool-age range (3-7 years old), having ongoing conventional physiotherapy and routine follow-ups in our clinic since birth. The exclusion criteria were having low birth weight or being premature, and having a systemic (e.g., diabetes mellitus, cystic fibrosis), genetic, or neuromuscular disease (e.g., Becker or Duchenne muscular dystrophy) or central nervous system disease (e.g., cerebral palsy), or having surgery in the last six months. However, the medical records of the participants showed that none of them had undergone any surgeries in the last eight months.

During the study period, 133 children with OBPP were screened during their routine controls; eight families refused to participate in the study. Nine patients were excluded because of surgery in the past six months, and four children were excluded, as the necessary data could not be collected. Therefore, the study included one hundred twelve children between the age of 44 and 77 (56.55 ± 8.69) months. Demographic descriptive data of the participants are presented in Table 1.

In order to determine and compare activity and participation levels of children with different injury types, we used the Narakas Classification System to classify the participants into three groups: Narakas Type 1 (n=7), Narakas Type 2 (n=91), and Narakas Type 3/4 (Narakas Type 3 n=2 and Narakas Type 4 n=12). Narakas Type 3 and 4 subjects were analyzed in the groups (n=14) since both types indicate total BP injury.

Based on the functions of the affected upper limb within the first two months after birth, the Narakas system classifies OBPP into four types, namely: Type 1 (C5-C6 root injuries), Type 2 (different severities of C5-C6-C7 root injuries), Type 3 (total lesion), and Type 4 (total lesion with Horner Syndrome) (4,20). The Narakas system could provide clinical data about the disease prognosis (20). A high rate of spontaneous recovery is commonly observed in Narakas Type 1 whereas, in total BP injuries (i.e. Narakas Type 3 and 4) spontaneous recovery is considerably less common (4,18,20).

We used the Pediatric Outcome Data Collection Instrument (PODCI) to determine activity and participation levels of the children. It is a parental report that evaluates activity, participation, and environmental factors in children with OBPP (12-14). Validated for OBPP, the PODCI consists of five subscales, namely, upper extremity functioning, transfer and basic mobility, sports and physical functioning, pain/comfort, and happiness (12,15). Both global score and subscale scores of the PODCI are evaluated over 100 standardized points. The Turkish version of the PODCI is a valid and reliable instrument to evaluate functionality in children and adolescents with cerebral palsy and chronic musculoskeletal disorders (21,22). We used the Turkish version of the PODCI with permission. The instrument and four of its subscales (upper extremity,

sports and physical functioning, pain/comfort, and happiness) were used in the study.

After collecting the participants' demographic data, the parents were informed about the PODCI. The instrument was delivered to the families in sealed envelopes and was collected back on the same day after the parents filled it out.

Statistical Analysis

Statistical analyses were performed using IBM SPSS version 23.0 (IBM SPSS Statistics for Windows, Version 23.0. Armonk, NY: IBM Corp). The assumption of the normal distribution of variables was examined using the Shapiro-Wilk test and histograms, boxplots, and Q-Q plot. At the same time, the descriptive statistics of the continuous data were given as mean and standard deviation for normally distributed variables, median (Quartile 1-Quartile 3) for non-normally distributed variables, and frequencies and percentages for categorical data. The Kruskal-Wallis variance analysis was performed to compare the PODCI scores of the different Narakas types since the normality assumption did not seem satisfied. Pairwise comparisons were conducted via the Dunn-Bonferroni test. A p-value of <0.05 was considered to be statistically significant (23).

The effect sizes are calculated by using "rcompanion" package in R to show the size of the difference between groups (24). The larger the effect size, the higher the degree that event will occur. The observed power is also given to present the statistical power of tests (25). Moreover, it was calculated using G*Power 3.1.9 (Franz Faul, Edgar Erdfelder, Albert-Georg Lang, and Axel Buchner, 2006) for parametric test version (One-Way ANOVA) and transformed to nonparametric test version by considering asymptotic relative efficiency of

Table 1: Characteristics of the Participants.

Characteristics		Narakas Type 1 (n=7)	Narakas Type 2 (n=91)	Narakas Type 3/4 (n=14)	p
		Mean±SD	Mean±SD	Mean±SD	
Age (months)		55.85±12.54	56.45±8.65	57.57±7.28	0.550
Gender, n (%)	Girl	2 (28.6)	6 (42.9)	51 (56.0)	0.577
	Boy	5 (71.4)	8 (57.1)	40 (44.0)	
Affected Side, n (%)	Right	6 (85.7)	10 (71.4)	61 (67.0)	0.241
	Left	1 (14.3)	4 (28.6)	30 (33.0)	

Kruskal-Wallis variance analysis (26, 27).

RESULTS

The children in Narakas Type 1 and Type 2 had similar and high scores on PODCI Global ($p < 0.001$), and subscales of Upper Extremity ($p < 0.001$), Pain/Comfort ($p < 0.001$) and Sport ($p < 0.001$). Both Groups' scores were higher when compared to Narakas Type 3/4. All of the effect sizes are greater than 0.26, which indicates a large effect size (25). The comparison of the PODCI global and subscale scores among the three groups is presented in Table 2 and Figure 1.

In the Happiness subscale of the PODCI, Narakas Type 1 had the highest scores, followed by Narakas Type 2 and Type 3/4 ($p < 0.001$). The comparison of the PODCI Happiness scores among the groups is presented in Table 2 and Figure 1.

DISCUSSION

We used the PODCI parental report to determine the activity and participation levels of preschool children with different injury types of BP. According to the results, the children in Narakas Type 1 and Type 2 (i.e., upper BP injury) had higher scores in PODCI Global, PODCI Upper Extremity, PODCI Pain/Comfort and PODCI Sport compared to the children in Narakas Type 3 (i.e., total BP injury). This finding indicated that compared to children with total BP injuries, those with injuries involving upper roots of the BP had higher levels of participation in activities of daily living and sports, and also more active use of their affected arm. The results also demonstrated that children with upper root injuries had less pain-related problems and more comfort during daily living than children with total BP injuries. Moreover, the children with the slightest in-

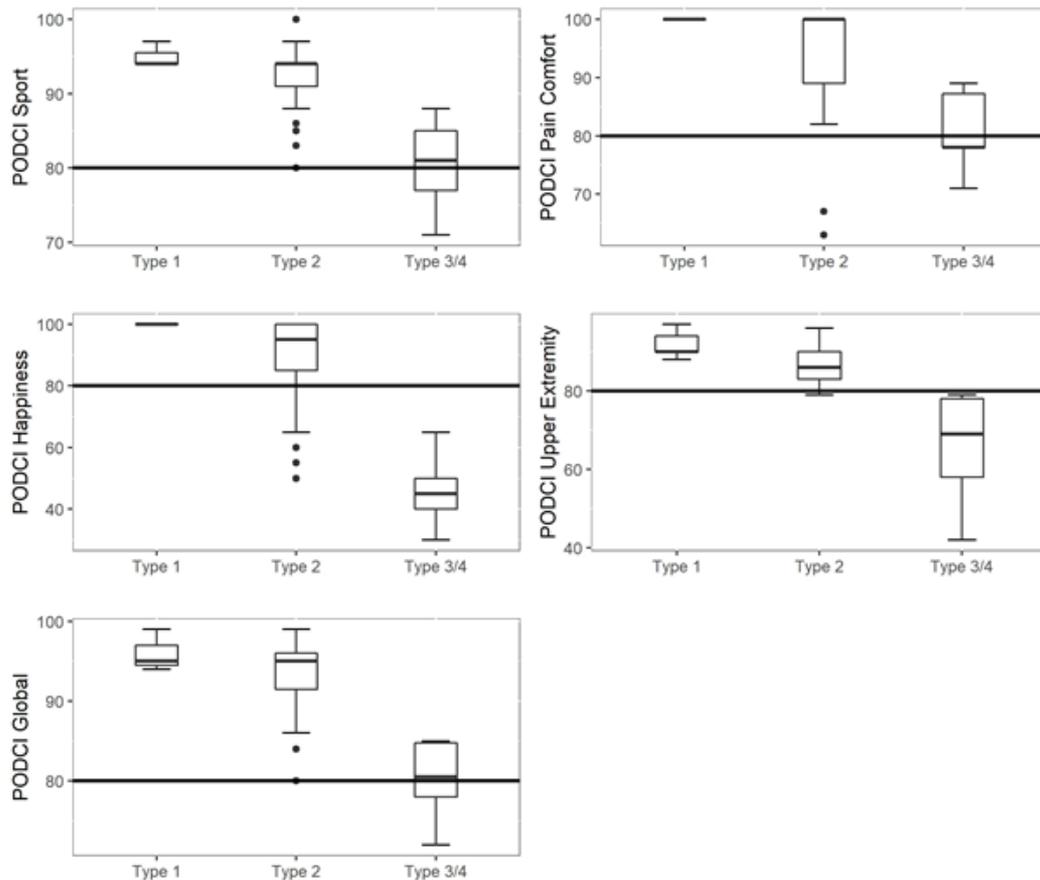


Figure 1: Box-plot Graphics. Results of the Pediatric Outcome Data Collection Instrument (PODCI) Global and Subscale scores (28, 29). Values below the thick black line (below 80 points) indicate functioning at a different level than the typical child, according to Haynes and Sullivan (30).

Table 2: Comparison of the Pediatric Outcome Data Collection Instrument Values among the Three Groups.

Pediatric Outcome Data Collection Instrument	Narakas Type 1 (n=7)	Narakas Type 2 (n=91)	Narakas Type 3+Tip 4 (n=14)	KW, ϵ^2 , Observed power	p
	Median (25-75)	Median (25-75)	Median (25-75)		
PODCI Sport	94.0 (94.0-97.0)	94.0 (91.0-94.0)	81.0 (76.25-85.0)	36.97, 0.333, 0.955	<0.001 ^{*b,c}
PODCI Pain Comfort	100.0 (100.0-100.0)	100.0 (89.0-100.0)	78.0 (78.0-89.0)	36.41, 0.328, 0.955	<0.001 ^{*b,c}
PODCI Happiness	100.0 (100.0-100.0)	95.0 (85.0-100.0)	45.0 (38.75-51.25)	43.60, 0.393, 0.955	<0.001 ^{*a,b,c}
PODCI Upper Extremity	90.0 (90.0-96.0)	86.0 (83.0-90.0)	69.0 (57.25-79.0)	41.05, 0.37, 0.955	<0.001 ^{*b,c}
PODCI Global	95.0 (94.0-99.0)	95.0 (91.0-96.0)	80.5 (77.5-85.0)	35.98, 0.324, 0.955	<0.001 ^{*b,c}

*p<0.05. 25-75: Percentiles. KW: Test statistic. ϵ^2 : Effect size. ^aStatistical Difference between Narakas Type 1 and Type 2 after Bonferroni Correction, ^bStatistical Difference between Narakas Type 1 and Narakas Type 3/4 after Bonferroni Correction, ^cStatistical Difference Between Narakas Type 2 and Type 3/4 after Bonferroni Correction. PODCI: Pediatric Outcome Data Collection Instrument.

juries (i.e., Narakas Type 1, C5-C6 injury) had the highest level of satisfaction regarding their activity performance and health status; followed by the children with Narakas Type 2 (C5-C6-C7 injury) and then the children with total BP injuries.

Many researchers have investigated recovery in BP in terms of joint movements or muscle strength and reported that functional deficits increase as the severity of injury increases (18,20). However, no studies are investigating activity and participation levels according to injury type or severity in OBPP. We investigated the activity and participation levels of preschool children with total BP injury and those with upper BP injuries. Our results indicated that different rates of spontaneous recovery in the post-natal period lead to various activity limitations and participation restrictions at older ages.

Preschool age range (3-7 years) was one of our inclusion criteria. Since OBPP requires long-term follow-ups, the focus of evaluation and treatment depends on the age. Evaluation and treatment are focusing on activity and participation gain importance as the children grow into preschool ages (13,31). As children grow into adolescents and adults, their activity and participation restrictions continue to persist or even worsen (11,32). It highlights the importance of detecting and addressing existing problems at preschool age.

Although few studies in OBPP have evaluated the activity and participation levels at different age ranges, none of these studies categorized the par-

ticipants according to their injury types (15-17,33). Most of the previous studies investigated whether children with OBPP had activity/participation problems or how their activity/participation levels differed from healthy controls. However, it is worth noticing that categorizing different injury types of OBPP as a single study group leads to heterogeneity in the functional capacity of the children. In 2008, Bae et al. showed that children with OBPP had lower scores than the normative data (15). However, the study does not provide information regarding different injury types that might be the source of these activity/participation restrictions. Most of the children in their study (64%) had early (severe) nerve injuries. Bae et al. in 2008 reported lower participation level of children with OBPP in sports compared to healthy children; whereas the same authors reported similar participation levels in 2009 (15,16). In another study published in 2009, Bae et al. evaluated 85 children with OBPP in the age range of 6-18 years, 19% of whom had Narakas Type 3 and Type 4 (children with total BP injuries) (16). According to the PODCI Sport scores, the children with OBPP had the same level of participation in sports as that of healthy children (16). According to our results, children with total BP injuries had lower participation in sports. The discrepancy between the studies may be due to the inclusion of two study groups included children with different injury types (15,16). Therefore, it is crucial to report the results of activity and participation evaluations in different types of BP injury.

Activity and participation could be evaluated using different scales and methods. Spaargaren et al. examined activity and participation in 53 seven- and eight-year-old children in a single group regardless of their injury type (17). Only three of the children included in the study had a total BP injury. Using parental reports, the investigators assessed the children's participation levels with various questions regarding their school performance, writing skills, bimanual activities, and leisure activities. The Children's Assessment of Participation and Enjoyment (CAPE) was also used to evaluate the children's participation in entertainment. According to the results, participants had mild problems in their writing skills. However, they had similar levels of participation in leisure and entertainment activities as their peers without any restrictions. In this study, the participants were not categorized based on their injury types, and the higher number of children with mild nerve injuries might have increased the participation level.

One of the critical findings in our study was that compared to children with upper BP injury, those with total injuries had a higher level of pain, leading to a lower level of comfort in daily life. Although it has been reported that children with OBPP have pain complaints (17,32), pain and comfort have not been previously investigated in different injury types of BP. Partridge and Edwards stated that there is an increase in the frequency of experiencing pain symptoms as the children grow (32). Studies investigating pain in OBPP are scarce, and our study contributes to filling this gap in the literature.

Regarding the Happiness subscale of PODCI, children with Narakas Type 1 (C5-C6 injury) were the group most satisfied with their activity performance, body and health status, followed by Narakas Type 2 (C5-C6-C7 injury) and Type 3 and 4 (total BP injury) who had relatively lower levels of satisfaction. Kirjavainen et al. studied satisfaction levels in children with OBPP who had undergone early nerve surgery (34). They reported an average of 6.1 points of satisfaction with appearance and 5.3 points of satisfaction with am-function on a Visual Analogue Scale ranging between 0-10. This finding indicates that children with low spontaneous recovery had lower levels of satisfaction that is a finding that is in line with our study results.

Our findings suggested that activity and participation levels differ depending on the type of BP injury. Haynes and Sullivan reported that scores below 80 on the PODCI could be clinically interpreted as an indicator of deviation from healthy normal children (30). Accordingly, in order to interpret our study findings, we added a thick black line in the boxplot graphs to highlight the 80-score limit representing healthy children. According to the graphs, children with Narakas Type 1 scored above 80, and there was a ceiling effect in some subscales. The scores of children with Narakas Type 2 fall within the range of 80-100; however, a few children scored below 80. These results indicated that the scores of all children with Narakas Type 1 and many children with Narakas Type 2 were within normal healthy limits. However, some children with Narakas Type 2 might have activity and participation restrictions. Majority of the children with total BP injuries (Narakas Type 3 and Type 4) scored below 80 in PODCI Global and its subscales. Considering the 80-point limit reported by Haynes and Sullivan, particularly children with total BP injury and few children with Narakas Type 2 may have activity and participation problems.

We observed that the scores of children with Narakas Type 1 and Type 2 had a ceiling effect in some subscales of the PODCI. Similarly, Dedini et al. shared the results of shoulder external rotation tendon transfer surgery and stated that the ceiling effect could be the reason for the lack of a relationship between postoperative improvements in the active range of movement and PODCI (35). The PODCI is recommended as one of the two scales that evaluate activity and participation in OBPP together with the self-care section of Pediatric Evaluation of Disability Inventory (PEDI) (12,14). However, since these are not OBPP-specific tools, they might fall short in evaluating activity and participation levels and detecting potential limitations. Although PODCI is commonly used in activity-participation studies in OBPP, the results of our study revealed a need for a more distinctive and disease-specific tool to evaluate activity-participation in OBPP, particularly for Narakas Type 1 and Type 2.

The unequal number of patients in the study groups was one of the limitations of the study. In partic-

ular, the low number of patients in Narakas Type 1 accounts for a statistical weakness. As reported in different sources, this group of OBPP patients has a recovery rate of 90-95% during the first year (2,18). Therefore, long-term medical follow-ups are not possible. To the best of our knowledge, there is no data in the literature about the activity and participation related to OBPP Type 1 (i.e., the slightest injury type) at preschool age. We included this group of patients (even though with a small number of participants) since we thought that this group might also have activity-participation restrictions, in contrast to the common belief of almost full recovery in these patients (2,18). Indeed, the scores of these children were not widely distributed, scattered or distant increases the reliability of the collected data. Due to the small number of patients in this group, we used nonparametric tests for group comparisons and calculated median and quarter values as descriptive statistics. Although the small sample size in Narakas Type 1 is the limitation of the study, we thought that the findings of the study were significant and could contribute to the literature. Because only 4-19% of all OBPP patients have a total injury (36), we thought that 14 patients in Narakas Type 3 (patients with total injuries) were adequate for this rare type of OBPP. Compared to similar studies, including patients with total injuries, our study had a higher number of participants. In several previous studies, children with Narakas Type 3 and Type 4 are included in one single group under the name of flail arm (37). Similarly, we also categorized all children with Narakas Type 3 and Type 4 in one group. However, the lack of investigation about Narakas Type 3 and Type 4, separately, was the second limitation of the study. Furthermore, there was not a healthy control group in our study. However, as recommended in the clinical use of the scale, in interpreting our findings, we used the score limit, which indicates deviation from normal healthy children.

In conclusion, preschool children with total BP injury were found to have lower activity and participation levels, less use of their arm, and lower level of participating in sports and daily life activities compared to children with upper BP injuries. Additionally, the scores of many children with total injuries were lower than the minimum limit of nor-

mal healthy children. Because activity and participation problems continue to exist or even worsen as the children grow, more emphasis should be given evaluating activity-participation and including relevant approaches in the treatment program of the children with total BP injuries. Our results also showed that children with total BP injury had pain complaints and lower comfort regarding their body. It also should be considered in medical follow-ups and treatments, and solution strategies should be developed. Although a small number of children with Narakas Type 2 had scores close to the lower score limit of normal healthy children, most of the children with Narakas Type 1 and Type 2 had high scores close to the ceiling score. It indicates the need for disease-specific and precise tools to evaluate activity and participation in this population.

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Informed Consent: A written informed consent form was obtained from the participants' families. All the families signed a declaration of informed consent.

Peer-Review: The authors will comply with the editor's decision on this matter.

Author Contributions: Concept – The research consists of a part of Kivanç Delioğlu's PhD thesis and investigates the activity and participation level of children with OBPP at preschool age; Design – Cross Sectional Study; Supervision – The supervisor of the PhD thesis is Mintaze Kerem Günel, PT, PhD, Prof. ; Resources and Financial Support – None; Materials – Facilities of Hacettepe University, Faculty of Physical Therapy and Rehabilitation were used by researchers. The Pediatric Outcome Data Collection Instrument was used in accordance evaluation. Data Collection and/or Processing –Narakas types of cases were determined by Akın Üzümcügil,

MD. As. Prof. and assessments in the content of the study were completed Kivanç Delioğlu, PT. MSc. Analysis and/or Interpretation – Statistical part of the study was analyzed by Ebru Öztürk, biostatistician. All authors contributed to interpret data. Literature Research – The literature was reviewed by Kivanç Delioğlu. Writing Manuscript – Kivanç Delioğlu wrote the first draft of the manuscript from his PhD thesis and all authors contributed in writing of manuscript. Critical Review – Revisions of the manuscript were completed by Mintaze Kerem Günel, PT, PhD. Prof and Kivanç Delioğlu together.

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